Chronic Melioidosis in a Patient with Cystic Fibrosis

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Burkholderia pseudomallei, the causative agent of melioidosis, is endemic in Southeast Asia and northern Australia, where it can be found in soil and surface water. We report a case of chronic pulmonary melioidosis in a patient with cystic fibrosis who had traveled to an area where B. pseudomallei is endemic.

CASE REPORT

The patient was a 38-year-old white male with cystic fibrosis (CF) who had undergone middle-lobe resection of the lung for bronchiectasis at the age of 17. During the following 10 years he has had only mild pulmonary symptoms. Since his lung function worsened during each winter season he started to spend the winter months in warmer areas: 1989 and 1990 in Indonesia and 1990 and 1991 in Mexico, the Dominican Republic, California, and Costa Rica. On returning from Thailand in the spring of 1992 he was severely ill with fever, cough, and malaise and was admitted to Freiburg University Hospital. The patient’s sputum sample yielded a gram-negative rod that was identified by API 20NE (code 1140474) as a Pseudomonas sp. Since then he had 13 documented exacerbations with gram-negative rods that were reported to be either Pseudomonas sp., Pseudomonas aeruginosa, or Burkholderia cepacia. In each case, he was treated for a maximum of 2 weeks with anti-pseudomonas antibiotics (ceftazidime, piperacillin-tazobactam, or piperacillin with or without aminoglycosides or ciprofloxacin) administered intravenously (i.v.) with only moderate effect. The isolated strains were susceptible to the drugs used with the exception of aminoglycosides. A detailed travel history was not documented, and B. pseudomallei and melioidosis were never suspected by the microbiologists and clinicians, respectively, associated with this case.

In November 1998 a new screening plate for Burkholderia species (3) had been implemented in the diagnostic laboratory, and a sputum isolate was sent to a reference laboratory (Munich, Germany). There, as in our own laboratory, Burkholderia pseudomallei was identified on the basis of biochemical tests (API 20NE code @48h 1156577) and by means of 16S rRNA gene sequencing and comparison to the type strain 1026b (Gen-Bank accession number U91839). Moreover, the isolate gave a positive reaction in a recently developed B. pseudomallei-specific latex agglutination test (11) which was not available in the diagnostic laboratory at the time of isolation. The isolate was susceptible to ceftazidime, imipenem, trimethoprim-sulfamethoxazole, tetracycline, and ampicillin-sulbactam and was resistant to all aminoglycosides, fosfomycin, and colistin. Bacte-
the API 20NE kit in order to get the correct identification has been reported (2), as well as misidentifications of *B. pseudomallei* as other species (4). A review of bacteriological reports in our case revealed that the API 20NE profiles of some former isolates were recorded after 24 h. Dance et al. have reported that 48 h of incubation of the panel is crucial for a correct identification of *B. pseudomallei* (2). We think it is reasonable to confirm a presumptive identification of *B. pseudomallei* based on biochemical results and resistance pattern by using serological methods such as the monoclonal antixopolysaccharide antibody-based *B. pseudomallei*-specific latex agglutination test (11). This might be especially true for laboratories where only occasional imported strains have to be identified. This case also demonstrates that testing patient sera for *B. pseudomallei*-specific antibodies might be a helpful tool for confirming the diagnosis.

Recently, imported melioidosis has been reported in patients with various underlying diseases (1), including one CF patient, but no history or clinical details were provided. It is highly suggestive that our patient became infected in Thailand, although we cannot completely rule out the possibility of a primary infection occurring during his visits to countries where *B. pseudomallei* is also endemic before he went to Thailand. Colonization and infection with *Burkholderia* species belonging to the *B. cepacia* complex and with *B. gladioli* is a well-recognized problem in CF patients and is associated with decreasing lung function and disease progression (7). One might speculate as to whether the changes in CF lungs predispose individuals to infection with *B. pseudomallei*. As a result of better management options for CF patients, the quality of life and life expectancy is increasing, and therefore patients’ travel activities may be increasing as well. Detailed history taking is crucial, and one should not be biased by the underlying disease when interpreting the biochemical results and susceptibility data. Awareness of melioidosis should be heightened since, as this case demonstrates, diagnosis may be delayed, therapy is difficult, and the outcome uncertain.

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REFERENCES